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CASE REPORT

Manifestation of carcinoma cuniculatum in the mandible

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Summary Carcinoma cuniculatum (CC) is a rare variant of squamous cell carcinoma usually occurring in the lower extremity. Occurrence in the oral cavity is unusual and was reported in the literature less than 20 times. A 81 year old female presented with pain in the left mandible. Clinical examination showed no pathologic result but panoramic radiograph showed osteolytic lesion. Result of histological study was CC and local resection was done. Eight month later patient presented again with pain in the contralateral side of the mandible and follow up X-ray showed a new osteolytic lesion. Histology confirmed second occurrence of CC. CC is rare entity especially in the head and neck showing local aggressive behavior but should be considered as differential diagnosis.

Introduction

Carcinoma cuniculatum (CC) is a rare disease first described Aird et al.[1] in 1954. It is a variant of carcinoma showing features of squamous cell carcinoma (SCC) and verrucous carcinoma (VC).[2] Histologically the tumour shows both, endophytic and exophytic growth, having papillomatous surface from which well-differentiated squamous cells penetrate deeply to the underlying tissue. The cellular elements are organized in ramified sinuses and crypts, which look like rabbit burrows and are full of ceratotic debris.[3] This typical characteristic is the origin of the name—cuniculus is the latin word for rabbit. CC predominantly affects older men but there are now definitive statements about etiology and pathology and only few data about recurrence rate or metastatic behavior exist.[4,5] A virological genesis is discussed because there are similar histological features seen in lesions caused by papillioma viruses. Due to its local aggressive behavior surgical excision with free margins is demanded as therapy of choice whereas the benefit of radio- and or chemotherapy is controversial. Typical localization of CC is the lower extremity especially

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the plantar region and some lesions off the sacral or genital region are described.[6] Involvement of the oral cavity is extremely rare, less than 20 cases are described in literature. Two intraosseous manifestations in one patient, especially in the mandible, not affecting the mucosa, was to the knowledge of the author not described before.

Case report

In August 2002 a 81-year old edentulous woman presented to our clinic and reported about ongoing pain in the anterior mandibular region on the left side over a period of several month. While clinical intra- and extraoral examination no

Figure 1 Panoramic radiographs demonstrates an unilocular radiolucent lesion in the left anterior mandible.

Figure 2 Radiological follow up after 8 months showing beside the defect resulting from further tumour enucleation new manifestation in the right anterior mandible.

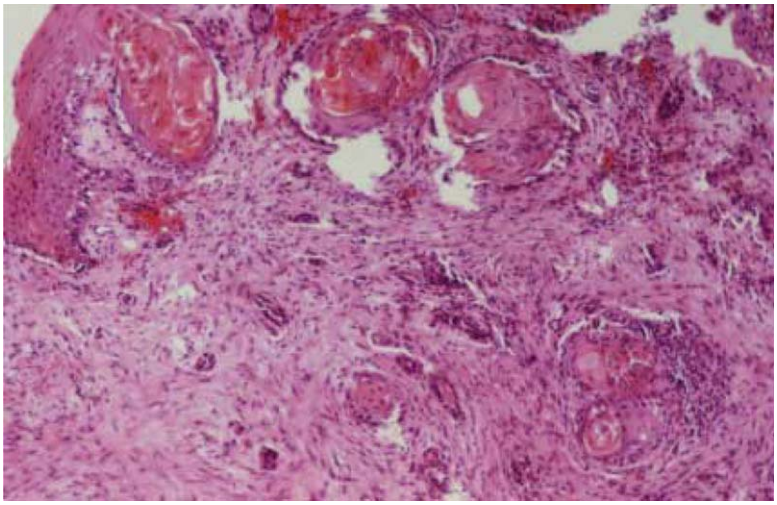

Figure 3 Histologic section (hematoxylin and eosin $\times$ 100) exhibits keratin filled crypts surrounded by well-differentiated squamous epithelium.

pathologic signs where found, panoramic radiographs demonstrated an unilocular radiolucent lesion in the anterior mandible (Fig. 1). Because of first appearance following extraction therapy osteomyelitis was diagnosed and patient was treated with antibiotics without any improvement of symptoms. Therefore, biopsy was taken under local anesthesia and histological examination showed diagnosis of well-differentiated squamous cell carcinoma. Due to reduced general condition (chronicle bronchitis, cardiac insufficiency, goat and thyroid complaint) we declined in arrangement with the patient a radical approach with mandibular resection and neck dissection but performed tumour enucleation with free margins in short general anesthesia. The pathologic finding was in contrast to first diagnosis on biopsy carcinoma cuniculatum, which was confirmed by a second reference pathologist.

Eight month later patient presented again with pain in the mandibular region but now on the contralateral side. While clinical examination again did not show any pathologic finding, panoramic X-ray showed beside the defect resulting from further tumour enucleation new lesion in the right mandible (Fig. 2). Intraoperative the region of first tumour occurrence was osseous well regenerated and biopsy remained negative. In the right mandible we found a new 2×2 cm osteolytic lesion with soft tumour mass which was resected. Pathologic finding confirmed the suspected second occurrence of CC. In a follow up period of 2 years, no new tumour or recurrence occurred.

Histopathology

Beside end differentiated squamous epithelium there are areas with better differentiation. There are layers with ortho- and parakeratotic hornformations looking like onion rings and many foci of microabscesses. The tumour exhibited only mild cytologic atypia but frequent mitosis and signs of lymphocyte infiltration was seen. All these features were indicative of carcinoma cuniculatum (Fig. 3).

Discussion

The pathogenesis of CC is unknown. Possible etiologic factors may be HPV, traumatic event, chronic inflammation, radiation or arsenic ingestion.[7] In oral CC alcohol and tobacco consumption were proposed in some reports as predisposing factor, but the real etiology remains unclear.[8,9] Patients are often in higher age and men seems to be predisposed. Intraoral presentation of the CC is typically mucosal exophytic lesion in the palatine area and the tumour is slow growing and local aggressive. Despite this, the here described case of an endophytic tumour involving the mandible in female patient without history of above estimated risk factors is absolutely uncommon and caused the first misdiagnosis of osteomyelitis. Furthermore in this case there were no pathologic signs in clinical examination and only by radiologic examination the tumour was obvious. One possible reason for the here described occurrence of CC may be an chronicle irritation of the mandible by prosthesis in edentulous situation. Although only less than 20 cases of oral CC have been reported to date, its frequency may be underestimated and misdiagnosed as VC or SCC. This is due to the difficult diagnosis on small amount of material typically are gained by biopsy. Even when in our patient after a follow up period of two years no recurrence was seen, the fact that a second primary occurred prompts the importance of short term recall.

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